Unusual presentation and treatment of spontaneous celiac artery dissection

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The management options of an isolated celiac artery dissection include medical, open surgical, and endovascular techniques. Which strategy is chosen depends on the severity of the dissection, collateral circulation to the liver, the patient's hemodynamic status, and the surgeon's expertise. We describe an unusual case of celiac artery dissection involving splenic and hepatic arteries complicated by hemorrhage. The patient was successfully treated by coil embolization of the splenic and gastric branches. Hepatic arterial blood flow was preserved with a stent graft extending from the origin of the gastroduodenal artery to the orifice of the celiac artery. (J Vasc Surg 2013;58:491-5.)

Isolated dissection of the celiac artery (IDCA) is very uncommon. Although the true incidence of IDCA is unknown, the widespread use of new diagnostic modalities has led to an increased number of reported cases in the last several years. While the development and application of new biomedical technologies have shed light on the pathophysiology of many vascular diseases, the etiology and natural history of IDCA has not been completely clarified.

There is also no consensus on the optimal management strategy for this disease entity. Most available case series suggest conservative medical therapy, but open surgical repair and endovascular repair have been applied on a case-by-case basis depending on the severity of the dissection and the patient's hemodynamic status.[1-6] The biggest challenge in using conventional endovascular interventions, such as coil embolization, is the potential risk of ischemic complications due to the difficulty in preserving flow through the branches of the celiac artery.[3]

We present a patient with IDCA extending into splenic and common hepatic arteries complicated by hemorrhage, which was repaired using endovascular techniques with complete exclusion of the celiac artery and preservation of the flow through the hepatic artery.

CASE REPORT

A 66-year-old woman presented to the emergency department with epigastric pain and nausea of 6 days in duration after hip replacement surgery 1 week earlier. Her medical history was noncontributory. She denied any history of trauma. A contrast-enhanced computed tomography (CT) scan of the abdomen revealed an isolated celiac artery dissection. The decision was made to manage the patient conservatively, and she was discharged on antiplatelet agents and given an outpatient vascular surgery clinic appointment.

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The patient presented to the emergency department several days later with nausea, vomiting, and worsening abdominal pain. She was normotensive, tachycardic, and afebrile. Laboratory testing revealed a drop in the hemoglobin to 6.7 from 9.3 g/dL 3 days earlier. Results of the liver function test and lactic acid levels were within normal reference ranges.

A CT scan showed dissection and aneurysmal dilatation of the celiac axis extending to the common hepatic and splenic arteries and free fluid around the pancreas and in the pelvis consistent with hemorrhage (Fig 1, *A* and *B*). The initial CT scan performed several days earlier did not reveal aneurysmal dilatation of the celiac artery or evidence of hemorrhage. The pancreas, aorta, and superior mesenteric artery appeared normal. No other cause of the intra-abdominal hemorrhage was found.

A decision was made to take the patient to our hybrid operating room to perform an angiogram with a view to endovascular intervention. The left common femoral artery was accessed, and a 4F introducer sheath was placed to allow quick placement of an aortic occlusion balloon in the event of significant bleeding with the manipulation of the celiac artery. Then, a cutdown, exposure, and catheterization of the left proximal brachial artery were carried out and an angiogram was performed. Upper abdominal aortography showed widely patent, normal-caliber superior mesenteric and renal arteries.

A selective angiogram of the celiac artery revealed a high-grade stenosis just distal to its origin, with aneurysmal dilatation of the more distal celiac artery extending to the common hepatic and splenic arteries (Fig 1, *C* and *D*). No active extravasation of contrast was seen. A superselective catheterization and angiography of the splenic artery revealed a long dissection. Six 8- × 7-cm and two 8- × 14-cm Nester coils (Cook, Bloomington, Ind) were placed into the splenic artery, successfully occluding the vessel (Fig 2, *A* and *B*). The left gastric artery was cannulated, and one 4- × 7-cm Nestor coil was placed near the origin of the left gastric artery that successfully occluded the artery (Fig 2, *C* and *D*).

An 8- × 5-cm Viabahn covered stent graft (W. L. Gore & Associates, Flagstaff, Ariz) was deployed just proximal to the origin of the gastroduodenal artery and extending to the orifice of the celiac axis. Completion angiography demonstrated good flow into the gastroduodenal and hepatic arteries, no flow into the aneurysm sac, and no visualization of the splenic or left gastric arteries (Fig 2, *E* and *F*). The left brachial arteriotomy was repaired with multiple interrupted 7-0 Prolene (Ethicon, Somerville, NJ)

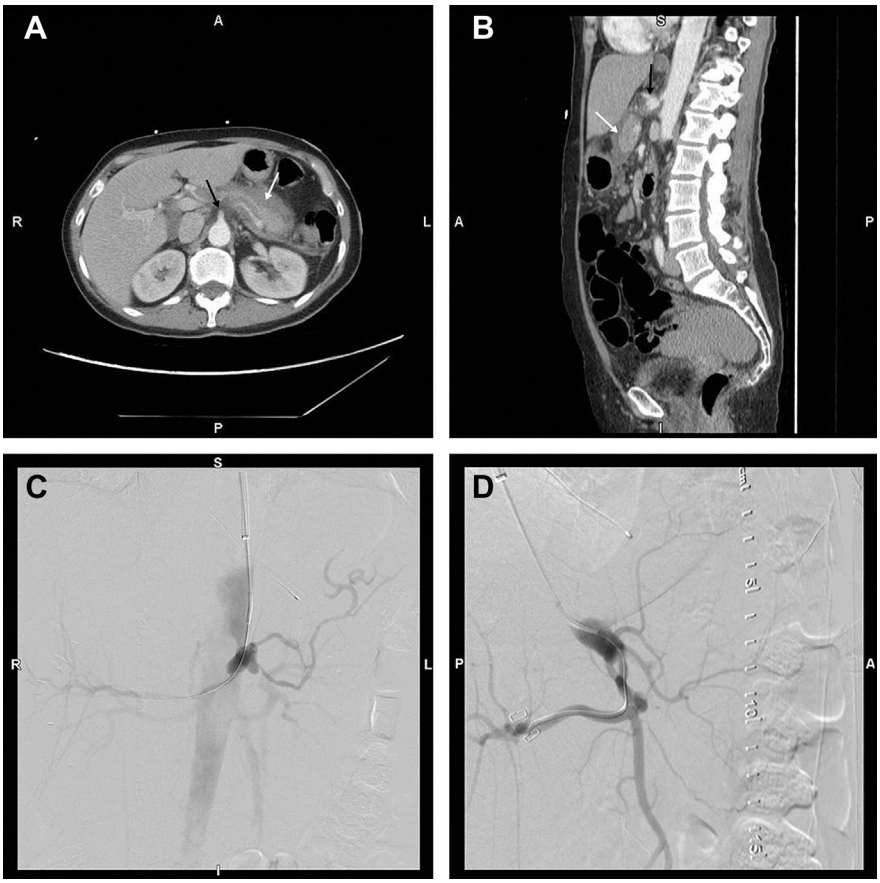

Fig 1. Preoperative computed tomography (CT) cross-sections in **(A)** axial and **(B)** sagittal views demonstrate the "double-lumen" sign (*black arrows*) of the dissecting celiac artery aneurysm with free fluid in the lesser sac (*white arrows*) consistent with hemorrhage. A celiac artery angiogram in **(C)** anterior and **(D)** lateral views demonstrates dissection and aneurysmal dilatation of the celiac artery extending to the common hepatic and splenic arteries.

sutures. The 4F sheath was removed from the left femoral artery, and pressure was applied while 50 mg of protamine was administered and hemostasis was achieved.

The patient's postoperative course was uneventful, and no increase was noted in serum transaminase levels. Blood serum tests performed for an autoimmune disorder suggested an atypical polyarteritis nodosa as a possible cause of the patient's condition. The patient was discharged 3 days later. On follow-up duplex ultrasound imaging performed 3 months postoperatively, the celiac-hepatic artery stent graft remained patent, and there was no evidence of endoleak or celiac artery aneurysm (Fig 3).

DISCUSSION

IDCA is rare, and only case reports are found in the literature. Between 1964 and 2011, 72 case reports were published, and 49 (68%) of these were reported in the last 5 years. Most of the patients were male (63 [88%]), and the average age at presentation was 54 years (range, 31-77 years).

IDCAs may be completely asymptomatic and were found incidentally in 14 patients (19%) surveyed for other conditions. When IDCA is symptomatic, the presentation is nonspecific. The most common presenting symptom is abdominal pain (51 [71%]), followed by back pain (six [8%]) and nausea (three [4%]). Chest pain (one [1%]), syncope (one [1%]), shock (one [1%]), and jaundice (one [1%]) have also been reported. Although no clear etiology is found in most patients, risk factors include hypertension, atherosclerotic disease, smoking, fibromuscular dysplasia, autoimmune conditions, trauma, infection, and connective tissue disorders.[4,6-8]

When IDCA is suspected, CT angiography (CTA) is the best diagnostic modality. In addition to being quick and accurate, it provides the most information in order to make therapeutic decisions. Magnetic resonance angiography is an alternative examination, but in our experience often does not provide the same detail as a CTA. In those patients in whom renal function is compromised, a color duplex ultrasound scan of the celiac axis may be diagnostic and an excellent test for follow-up evaluation. It does not, however, provide the necessary detail to make treatment decisions. Intravascular ultrasound imaging may aid intraoperatively in identifying the true vs false lumen and perhaps the site of rupture. The primary disadvantages of

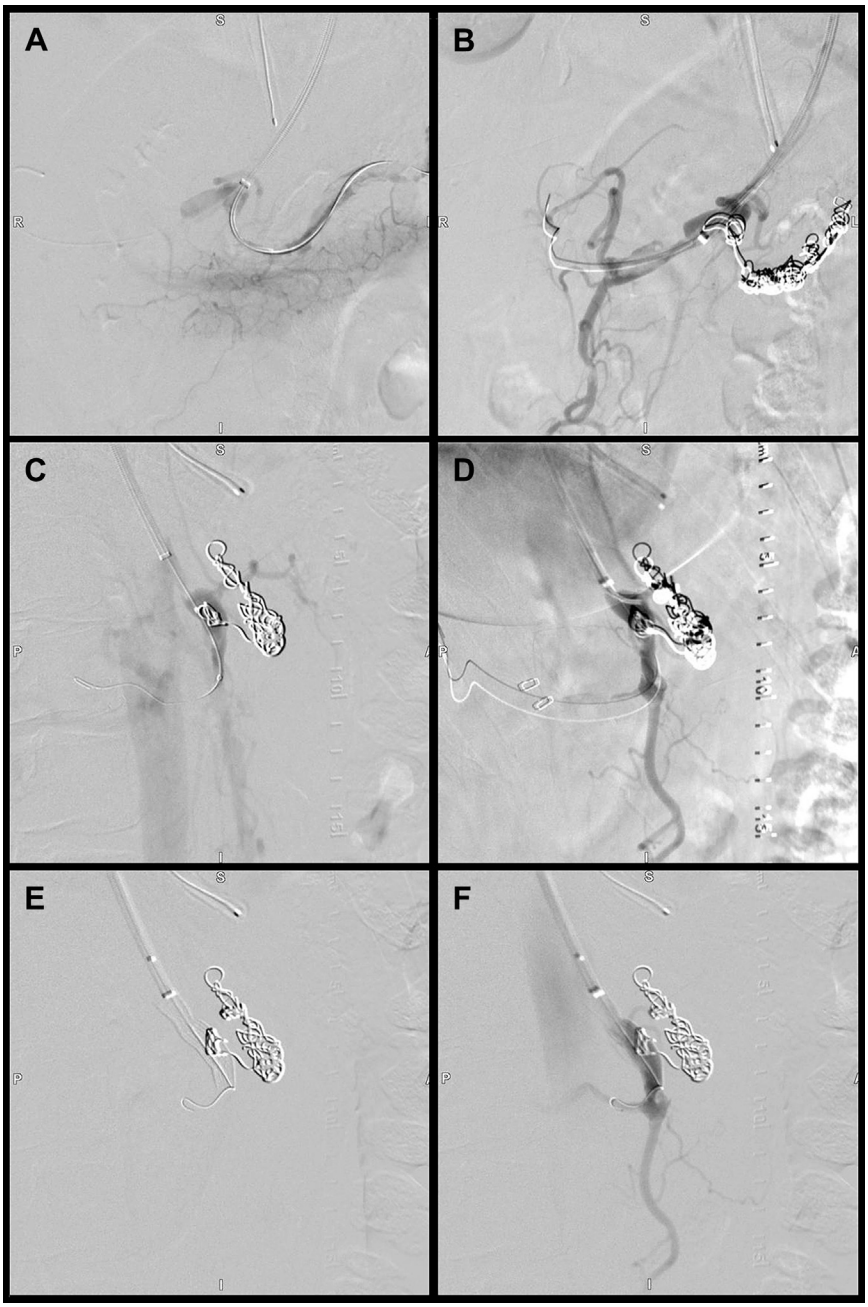

Fig 2. An angiogram of the splenic artery reveals **(A)** aneurysmal dilatation and long dissection of the artery and **(B)** coil embolization. **C,** Shows angiogram of the left gastric artery visualized posterior to coiled splenic artery, **(D)** single-coil embolization of the left gastric artery, and **(E)** deployment of the stent graft just proximal to the origin of the gastroduodenal artery and extending to the orifice of the celiac artery. **F,** A completion angiogram demonstrates good flow into the gastroduodenal and hepatic arteries and no flow into the aneurysm sac.

intravascular ultrasound imaging, however, are its expense and the significant increase in the operative time.

The management of IDCA aims at preventing the expansion of the false lumen, which may cause malperfusion, aneurysmal dilatation, or rupture. Liver function tests may be helpful in determining if there is hepatic malperfusion and ischemia that might prompt more aggressive management. The literature shows that in the absence of malperfusion, conservative management with antihypertensive and anticoagulation therapy was the most common treatment modality (45%-63%).[4,5,9-23] Most of the patients treated conservatively were discharged when their symptoms resolved and were

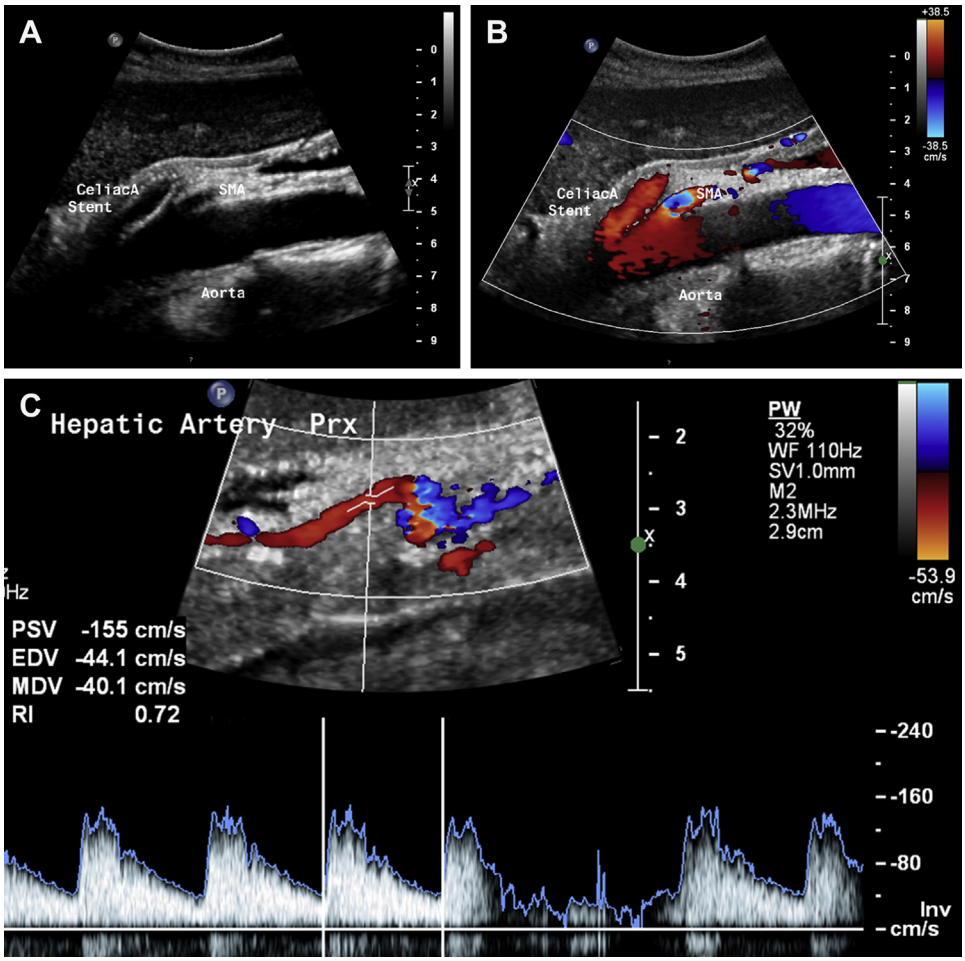

Fig 3. A follow-up duplex ultrasound image at 3 months demonstrates **(A)** celiac-hepatic artery stent graft, **(B)** continued aneurysm exclusion with no evidence of endoleak, **(C)** patent hepatic and gastroduodenal arteries. The ultrasound cursor is in the common hepatic artery.

followed up with CTA within 1 week to 3 months.[4,9,16] Although there is no consensus on the duration of conservative therapy, treatment with anticoagulant or antiplatelet agents from 3 weeks to 6 months has been reported to achieve good outcomes.[4,16,21] In those patients treated with warfarin, an international normalized ratio target of 2.0 to 3.0 is sought. In addition, strict blood pressure control was also seen as an important factor in stabilizing the celiac artery dissection.

Findings in 25 (35%) of the reported patients with IDCA were consistent with liver malperfusion on the initial CTA or propagation of the dissection on follow-up CTA and worsening or recurrent symptoms. In these patients, open surgery or endovascular intervention was undertaken. Nineteen patients (76%) underwent open procedures. Of those, 14 had a vascular bypass to the hepatic artery using prosthetic or great saphenous vein graft, three patients had resection of the celiac artery and direct anastomosis of the hepatic artery to the ostium of the celiac artery, and two patients underwent ligation of the dissected celiac artery without revascularization. Endovascular intervention was performed in six patients (24%). Three of these patients (50%) had coil embolization of the celiac artery and the hepatic, splenic, and gastric artery branches.[2,6,24] Stenting of the celiac artery was performed in three patients (50%).[3,7,25] One patient had a combined procedure with coil embolization of the celiac artery and an open hepatic artery bypass.[6]

Bleeding or rupture is a life-threatening complication of IDCA. Most ruptured IDCAs resulted in sudden death before any treatment and were diagnosed at postmortem examination.[26,27] One patient in hemorrhagic shock survived after coil embolization of all arterial branches of the celiac artery.[2]

Collateral blood flow to the liver through a patent gastroduodenal artery should minimize the risk of hepatic ischemia with coil embolization of the common hepatic artery. Nevertheless, some risk of hepatic ischemia and celiac aneurysm late rupture still exists with this method of treatment. We were able to successfully treat our patient by coil embolization of the splenic and gastric branches with

preservation of the hepatic arterial blood flow and aneurysm exclusion by deploying a covered stent graft extending from the orifice of the celiac artery to the origin of the gastroduodenal artery. To our knowledge, this is the first published report of such a procedure performed for the management of an IDCA complicated by aneurysmal dilatation and hemorrhage.

It is difficult to argue that preservation of the hepatic artery would not be preferred to coil embolization when technically possible and if there is no significant additional risk or procedure time. Furthermore, continued aneurysm growth and rupture of visceral artery aneurysms treated with coil embolization only has been reported.[28-30]

CONCLUSIONS

Although determining the long-term durability of endograft techniques will require further studies, stent graft repair of visceral artery aneurysms shows an early promise as a safe and effective treatment by excluding the aneurysm and minimizing the risk of aneurysm expansion or rupture.

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